

Management of Paediatric Oral Ranula with review of literature: A case report during Covid-19 pandemic

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ABSTRACT

Ranula is a term used to describe a soft, clear or bluish swelling that usually appear in the floor of mouth under the tongue. Ranulas are classified as simple, or plunging. Simple ranula occurs in the floor of the mouth as bluish coloured swelling while plunging ranula extends deeply into the submandibular and neck spaces to manifest clinically as a cervical swelling after herniation of the pseudocyst through the mylohyoid muscle. The present case reported a 5- year-old female patient with oral ranula. The classical clinical features of oral ranula as well as the surgical management of the case has been described in details. The case report also presents a comprehensive discussion of the recent literature on the oral ranula.

التعامل مع الكيس اللعابي الفموي عند الأطفال مع مراجعة ماكتب في السابق بخصوص هذا الموضوع: تقرير حالة أثناء جائحة Covid-19

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الكلمات المفتاحية:

كوفيد-19
الإدارة
الفم
الأطفال
رانولا

الملخص

رانولا مصطلح يستخدم لوصف تورم رخو واضح أو مزرق يظهر عادة في أرضية الفم تحت اللسان. يتم تصنيف Ranulas على أنها بسيطة، أو غاطسة. تظهر الرانولا البسيطة في أرضية الفم حيث يمتد التورم الملون المزرق أثناء غرق الرانولا بعمق في الفراغات تحت الفك السفلي والرقبة لتظهر سريريًا على شكل تورم في العنق بعد فتح الكيس الكاذب من خلال العضلة النخاعية. الحالة الحالية مريضة تبلغ من العمر 5 سنوات مصابة بكيس لعابي فموي. تم وصف السمات السريرية الكلاسيكية للرانولا الفموية بالإضافة إلى الإدارة الجراحية للحالة بالتفصيل. يقدم تقرير الحالة أيضًا مناقشة شاملة للمراجع الحديثة حول الرانولا الفموية.

Introduction

The term ranula, used to describe a bluish translucent cyst in the floor of the mouth, it is derived from the Latin word "rana" that denotes the shape of the abdomen of a frog [1]. In dental literature, two main theories have explained the mechanism of ranula growth. The first theory, proposes that extravasation of mucus results in development of ranula, while the second theory explain that ranulas occur as a

result of mucus retention [2]. The precise data about the incidence rate of ranula is uncertain, however, some researches have described it as 2/10000 [3] and associated with propensity toward female than male with a male-to-female ratio was reported as 1:1.4 [4]. The vast majority of case reports showed that the incidence of intraoral ranula is highest among patients in the 2nd and 3rd decades of life [5-8], in

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addition, the lesion can affect patients in their 1st decade of age [9]. However, it has been reported in literature that ranulas occurred in patients with ages ranging from 3 years to 61 years [1]. It has been proved in dental literature that a large number of age group were developed intra oral ranulas as youngers of three months to up of 80 years [5]. The present case reported a case of oral ranula involving floor of the mouth in a 5-year-old female patient with comprehensive management and surgical treatment. The importance of this case report lies on the following points: firstly, the relatively large size of the ranula in young age group of 5-year-old child. Secondly, the challenging in the diagnosis and management of such case during the Corona virus (Covid-19) pandemic where access to hospital medical services and facilities in Libya is critical at this time.

Case report

A 5-year-old Libyan female referred to the maxillofacial unit at National Cancer Institute of Misrata city, accompanied with her parent, complaining of a swelling under the tongue progressed gradually to considerable size in relatively short period of time of less than one day. The parent reported that her daughter complained many times of discomfort originating from the area under the tongue but was not impairs the swallowing, speech, chewing or breathing. The girl was otherwise in a good health, active, cooperative with no history of any recent nausea, vomiting, fever or weight loss.

Clinically the patient appeared fit and well with no history of oral trauma. On intra oral examination, the floor of the mouth enlarged on the right side with normal overlying mucous membrane. The resultant swelling was smooth, hemispherical in shape occupying the whole right-side space of the floor of the mouth with a tendency to cross the midline, pushing the lingual frenum toward the left side. An obvious blood capillaries were visible on the lesion surface without any sign of discharge [Figure 1]. On palpation the swelling was not tender or painful, firm in consistency, approximately 3 centimetres in diameter. The patient's oral cavity including oral mucus membrane, gingiva, palatal surface and dentition all appeared normal with good oral hygiene.

On extra oral examination, no cervical lymphadenopathy was present, and the patient's neck was supple with no indication of any cervical swelling.

A provisional diagnosis of ranula has been made on the basis of history and clinical features. Non-invasive diagnostic method was taken for diagnostic purpose, such as a neck ultrasound with focusing on the floor of the mouth and submandibular area. Neck ultrasound confirmed the diagnosis of ranula with no indication of any calcific duct obstruction [Figure 2]. In the second visit, after three days, the patient's father mentioned that the swelling's colour has changed to blue and became associated with moderate pain while chewing food and speaking, the thing that forced his daughter to stop eating solid foods and restricted her speech.



Figure 1. An oral ranula on the floor of the mouth/ right side, causing deviation of the lingual frenulum toward the left side and crossing the midline.

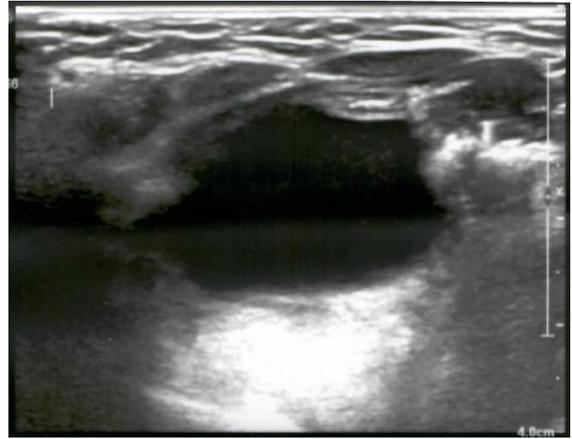


Figure 2. Pre-operative ultrasonographic image, demonstrating the isolated ranula.

It appears as a hypoechoic homogeneous cystic lesion that has a well-confined boundary and measured 2.5 x 1.4 x 2.3 cm.

As the lesion become larger in size of (more than 2 cm), a CT scan of the neck area has been done to rule out the possibility of plunging ranula formation although there was no clinical sign of neck involvement as an indicator of that. The axial CT scan study of the floor of the mouth and upper neck with 30 ml intravenous (I.V) contrast in 3 mm slice thickness revealed presence of 41.5 x 14.5x 35 mm thin wall oval cystic lesion with fluid content density of sublingual space. It had mild compression on the base of the tongue and subsequently on the oropharynx as well. It does not extend down to the submandibular space and not communicating to the adjacent structures [Figure 3].

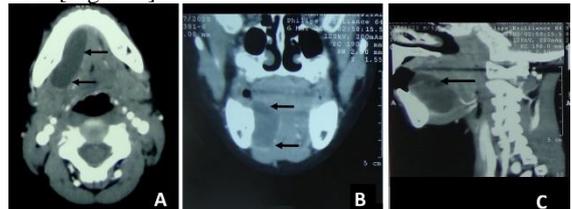


Figure 3. Contrast-enhanced CT scans: (A) axial and (B) coronal show elongated cystic lesion in right sublingual space (arrows) and (C) sagittal view shows well visualized cystic lesion.

After the lesion confirmed as a ranula, the treatment planned as marsupialisation accompanied by surgical removal of related sublingual salivary gland under general anaesthesia. Preoperative assessment and investigation carried out. Nasal intubation performed, the operating theatre was disinfected followed by aspiration of cyst's content that yielded a clear fluid, confirming that no infection or pus in the lesion. The aspirated liquid appeared to be highly viscous in nature. A two cm incision was performed over the surface of the swelling only on the mucosa which was directly bounded by the capsule of the mass. All cystic content was completely evacuated from cystic cavity. Surgical stitching for purpose of marsupialisation was carried out using 3-0 sutures (DAMACRYL: Synthetic Absorbable Surgical Suture, Polyglycolic Acid, Coated, Braided by GMD Group Medikal San. Tic. AŞ) which insuring suturing the lesion lining membrane with the floor of the mouth mucosa to evade recurrence. The related sublingual salivary gland was found and surgically removed too [Figure 4]. The surgical cavity was filled with iodoform gauze as a local antiseptic and sutured. An excisional biopsy from Cystic tissue was sent for histopathology. The following medications were prescribed to the patient during her admission in the hospital for two days:

Intravenous administration of Augmentin 300mg/12 hours, Flagyl 125mg / 12 hours, Dexamethasone 4mg/ 12 hours and Perfelgan 250mg/12 hours. The antibiotics were instructed to be continued for another 3 days and to undergo a soft diet regimen. Two days Post-operative follow up was in the hospital under direct clinician's observation. The surgical field was inspected in dental clinic as the follow: in the next day, three days later the iodoform dressing was removed followed by copious irrigation with saline and one week later, healing was found favorable. Due to the pandemic of Covid-19

and the associated restrictions applied by the authorities regarding the travel between the cities in order to control spread of the disease, the treating team continued to follow-up the patient from a distance over phone contact. For a period of more than three months no sign or symptoms of relapse was reported by the parents of the child. The histopathological analysis of the cystic lesion confirmed that the growth was ranula.

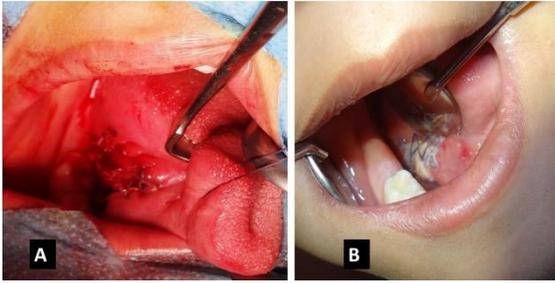


Figure 4. A. Suturing the lesion lining membrane with the floor of the mouth mucosa. B. Iodoform packing used to fill the ranula cavity. The gauze were sutured to surgical site.

Discussion

Ranula is essentially a mucocele that implicates the sublingual salivary gland and occurs as a result of the salivary extravasation of any of the 20 ducts of the sublingual salivary gland [10]. On top of the two proposed theories for oral ranula aetiopathogenesis, other factors have also been suggested. Increasing incidence of ranula has been reported in certain ethnic groups such as Maori and Pacific Island Polynesians, and this signifies the possibility of congenital factors contribution in the development of ranula [11]. It has been reported that oral ranulas manifest in the floor of the mouth as a blue, dome-like swelling with size of approximately 3-6 centimeters [10]. Further expansion of the swelling may result in dysphagia, difficulty in speech, or even airway blockage [5, 8]. In pediatric patients under the age of five years, an untreated ranula may lead to obstructive sleep apnea, and delay in seeking treatment can eventually result in growth disturbance [6].

In general, ranulas are categorized as simple and plunging types based on their spread [12]. Typically, plunging ranulas are known to have posterior spread further than the free edge of the mylohyoid muscle encroaching the sub- mandibular and sometimes even the parapharyngeal spaces [12-14]. Majority of the described cases revealed that, lateral side of floor of the mouth is the common site of ranulas [13]. In rare cases, they may progress to involve both sides and present as a bilateral swelling [5, 7]. Minor gender preference toward the female population was stated in reviews although the difference in this predilection was not substantial [15]. In keeping with previously published data, our patient was a female with occurrence site was found in the lateral right side of the mouth floor. Furthermore, the observed clinical features of the ranula were parallel to related literature and its size was roughly 3 centimeters. However, the age of our case was 5 year and this was in contrast to what has been reported by several other case report in literature with highest incidence being reported in the second and third decade of life [14-16].

Lesions well known to occur in the floor of the mouth such as lipoma, dermoid cyst, abscess, salivary gland lesions and vascular lesions are considered to be differential diagnosis for oral ranula with no particular diagnostic tests to diagnose ranula [10]. In most of the cases simple ranula manifests as a cystic fluctuant growth which characterises with progressive increase in size. It has been reported that biochemical analysis of ranula contents showed presence of elevated salivary amylase and protein concentration in comparison with serum hence suggestive of ranula arise out of sublingual gland which delivers saliva with increased protein levels when compared with submandibular gland [17].

Generally, ranula diagnosed from its classic clinical features; however, several methods have been used, to investigate ranula and differentiated it from other oral soft tissue lesions. It is ranging from invasive techniques such as Fine Needle Aspiration Cytology (FNAC) to non-invasive imaging procedures such as dental radiographic occlusal view and Magnetic Resonance Imaging (MRI)

[16]. Fine-needle aspiration of the lesion can help diagnose ranula smaller than 2 cm, and will show straw-coloured aspirate. Furthermore, the presence of granulation tissue and a chronic inflammatory reaction will be demonstrated by histopathological examination [5]. Ultrasonography (US) is a useful imaging method for the sublingual space, particularly for simple ranulas, as it is unaffected by metal dental restoration such as amalgam and can locate the lesion [18]. Furthermore, ultrasonography has been suggested as a key component in the management of floor-of-the-mouth masses in children [19]. US with high resolution is preferred over other imaging techniques as it is easily accessible, does not implicate radiation and does not necessitate sedation. Moreover, an ultrasound is able to show sublingual gland herniation and aid in defining the degree of the pseudocyst's involvement in the cervical space for a plunging ranula [20]. On US images, a ranula typically appears as a homogeneous cystic (anechoic) lesion deep to the mylohyoid muscle that has a well-confined boundary [21]. It may contain fine internal echoes due to debris [22]. In the present case ultrasound was made three days after initial diagnosis. Ultrasound report stated presence of evidence of hypoechoic cystic lesion measured 2.5 x 1.4 x 2.3 cm seen at right side of floor of the tongue. In more progressed cases, if the ranula is exceeding 2 cm, an MRI or CT scan is recommended to determine size and extension of the lesion [8]. In plunging ranulas, a CT scan image will show a classical tail sign extending from the sublingual gland through the mylohyoid muscle, which differentiates it from other pathologies [20]. A number of techniques have been used for management of oral ranula which includes sclerotherapy, marsupialization, surgical removal of the pseudocyst, and surgical removal of the sublingual gland. [23]. The more prevalent encouraged procedure for the treatment of ranula in the past was marsupialization. It is associated with high recurrence rate ranging between 61% and 89% as the pseudocyst was not entirely eradicated. In some cases, it acted as an originator for plunging ranula. The packing of the ranula cavity with gauze after marsupialization was described to be useful in decreasing the rate of recurrence. It is commonly appropriate for ranula with less than 2 cm in diameter [8, 24]. It has been recommended to excise the cyst along with the related sublingual gland in order to avoid this type of complication [8, 25]. The rate of recurrence of ranula varies according to the method of treatment used. The highest recurrence rate, ranges from 70% to 100%, was reported when ranulas treated with incision and drainage whereas ranulas treated with marsupialization registered a recurrence rate ranges from 61%-89% (also other researches mentioned a range from 36.4% to 80%) [26]. The lowest recurrence rate, ranges from 0% to 3.8%, was found for ranulas treated with excision of ranula along with sublingual salivary gland [26], however, for ranulas treated with surgical excision only without removing the sublingual salivary gland have recurrence rate ranges from 18.7% to 85% [26]. According to the previous mentioned factors and elements, we preferred to manage the present case with marsupialization of the cyst along with surgical excision of sublingual salivary gland. The marsupialization procedure was modified by packing the cystic cavity with iodoform gauze.

Furthermore, another factor that had a strong influence on our selection for this type of management, beside patient's age and size of the ranula, is the general condition of the health services in Libya during the pandemic of Corona virus (Covid-19). As many surgical procedures in public and private practice has been limited to very restricted emergency cases only.

Surgical management of ranulas can be associated with a number of commonly described complications such as recurrence of the lesion post-operative bleeding or hematoma, wound dehiscence and infection [23, 26]. Surgical removal of the sublingual salivary gland is linked with a main risk of injury to the lingual nerve leading to sensory deficit of the tongue, and damage of the Wharton duct [5, 23]. Although size of ranula in the present case was relatively large, no complications were observed during the week after surgery and no unfavorable events as an evidence of recurrence was reported by her parents over phone for a period of three-month post-operatively.

Conclusion

A key element in the diagnosis of ranula is the identification of remarkable clinical features of ranula. Different methods can be used

to investigate ranulas and determine its type particularly in cases of plunging ranula, or to rule out differential diagnosis. Packing of the ranula cavity with gauze after marsupialization was described to be useful in decreasing the rate of recurrence. It has been recommended to excise the ranula along with the related sublingual gland in order to avoid a complication of recurrence.

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